Research Article

Executive Functions Impact the Relation Between Respiratory Sinus Arrhythmia and Frequency of Stuttering in Young Children Who Do and Do Not Stutter

Robin M. Jones,^a Tedra A. Walden,^b Edward G. Conture,^a Aysu Erdemir,^b Warren E. Lambert,^b and Stephen W. Porges^c

Purpose: This study sought to determine whether respiratory sinus arrhythmia (RSA) and executive functions are associated with stuttered speech disfluencies of young children who do (CWS) and do not stutter (CWNS).

Method: Thirty-six young CWS and 36 CWNS were exposed to neutral, negative, and positive emotion-inducing video clips, followed by their participation in speaking tasks. During the neutral video, we measured baseline RSA, a physiological index of emotion regulation, and during video viewing and speaking, we measured RSA change from baseline, a physiological index of regulatory responses during challenge. Participants' caregivers completed the Children's Behavior Questionnaire from which a composite score of the inhibitory control

and attentional focusing subscales served to index executive functioning.

Results: For both CWS and CWNS, greater decrease of RSA during both video viewing and speaking was associated with more stuttering. During speaking, CWS with lower executive functioning exhibited a negative association between RSA change and stuttering; conversely, CWNS with higher executive functioning exhibited a negative association between RSA change and stuttering.

Conclusion: Findings suggest that decreased RSA during video viewing and speaking is associated with increased stuttering and young CWS differ from CWNS in terms of how their executive functions moderate the relation between RSA change and stuttered disfluencies.

In Memoriam: Dr. Edward "Warren" Lambert, 1945-2017

We happily look back on our many walks under Vanderbilt University's magnolia trees to the Vanderbilt Kennedy Center to meet on research and statistical issues with Dr. Edward "Warren" Lambert. For past and present members of Vanderbilt's Developmental Stuttering Project, Warren was a trusted colleague, friend, and statistical consultant whose wit and wry sense of humor always made us smile and helped ease the challenging realities of the analytical work in front of us. Dr. Lambert left us on January 13, 2017, but he will long be remembered and not quickly forgotten by his Vanderbilt friends, colleagues, and students. Truly, Warren

was a friend, a confidant, a gentleman, a scholar, the epitome of a wonderful human being, and a significant contributor to our work. However, it wasn't just the amount of help he gave us, but the way in which he gave it. We are all the better for learning from, knowing, working with, and laughing with him.

We are positive that Warren has gone to that place where all good mentors, scholars, and researchers go. We will miss him tremendously. Warren, rest in peace. You have more than deserved it.

espite advances in the understanding of the neural, speech-motor, linguistic, and emotional contributions to the etiology of childhood stuttering, several questions remain. One question relates to the fact that stuttering is quite variable; for example, it varies within and between speaking situations (e.g., Johnson, Karrass, Conture, & Walden, 2009), with the precise reason for such variation unclear. Thus, empirical study of factors that influence stuttering both between and within speaking situations for young children should serve to advance our

Correspondence to Robin M. Jones: robin.m.jones@vanderbilt.edu

Editor: Julie Liss

Associate Editor: Hans-Georg Bosshardt

Received March 21, 2016 Revision received August 3, 2016 Accepted February 8, 2017

https://doi.org/10.1044/2017_JSLHR-S-16-0113

Disclosure: The authors have declared that no competing interests existed at the time of publication.

^aVanderbilt University School of Medicine, Nashville, TN

^bVanderbilt University, Nashville, TN

^cIndiana University Bloomington

understanding of contributors to the onset, maintenance, and/or exacerbation of stuttering. Emotional reactivity and regulation represent processes that vary in response to the changing affective/social contexts of various communicative situations and may provide new insights into the variability of stuttering.

Emotion and Stuttering

Recently, several empirical investigations have studied differences in temperament and emotions between children who stutter (CWS) and children who do not stutter (CWNS), as well as the impact of emotional processes on stuttering frequency (e.g., Eggers, De Nil, & Van den Bergh, 2010; Jones, Buhr, et al., 2014; Karrass et al., 2006; Walden et al., 2012). Reviews of these studies (Conture, Kelly, & Walden, 2013; Jones, Choi, Conture, & Walden, 2014; Kefalianos, Onslow, Block, Menzies, & Reilly, 2012) suggest that there is an association between aspects of emotion and childhood stuttering. For example, CWS, compared to CWNS, exhibit: (a) less adaptability (e.g., Anderson, Pellowski, Conture, & Kelly, 2003; Schwenk, Conture, & Walden, 2007) and (b) more negative affect/mood (e.g., Jones, Conture, & Walden, 2014; Ntourou, Conture, & Walden, 2013). In addition, increases in CWS's stuttering have been associated with (a) higher emotional reactivity and lower emotion regulation (e.g., Arnold, Conture, Key, & Walden, 2011; Jones, Conture, et al., 2014; Kraft, Ambrose, & Chon, 2014; Walden et al., 2012) and (b) emotionally arousing conditions (e.g., Choi, Conture, Walden, Lambert, & Tumanova, 2013; Johnson, Walden, Conture, & Karrass, 2010). In contrast, a recent review (Alm, 2014) suggested that preschool-age CWS as a group were not more likely to show temperamental traits of shyness or social anxiety than CWNS; however, this review also suggested that CWS exhibited traits associated with inattention and hyperactivity/ impulsivity. Findings to date suggest that this relation deserves continued empirical study.

When studying the association of emotion and childhood stuttering, several methodological approaches may be considered. One approach—psychophysiology—is attractive because it involves a relatively objective means to study emotional processes. To date, physiological measures (e.g., measurement of autonomic nervous system [ANS] activity) have received little empirical attention in the study of young CWS. This is unfortunate, since ANS activity provides insights into covert processes of emotional reactivity and regulation and thus augments the larger body of evidence based on more overt expressions of emotion (e.g., caregiver report and behavioral observation). Further, the importance of using multiple measures in studying emotion should be underscored. While there is no unitary consensus on what constitutes emotion, there is a strong agreement among experts that one important and defining feature of emotion is physiological activity (Cabanac, 2002; Izard, 2010; Kreibig, 2010; Sonnemans & Frijda, 1994). Thus, in order to establish a comprehensive understanding of the relation between emotion and stuttering, physiology should be included (for

a more in-depth perspective on the authors' use of physiology as an index of emotion in association with childhood stuttering, see Jones, Buhr, et al., 2014).

There is a limited but growing number of empirical studies that have examined the ANS activity of young CWS and CWNS (Choi, Conture, Walden, Jones, & Kim, 2016; Jones, Buhr, et al., 2014; Zengin-Bolatkale, Conture, & Walden, 2015). Notable findings from these studies have indicated that: (a) CWS exhibited lower parasympathetic activity (index of emotion regulation) at baseline and greater sympathetic activity (index of emotional reactivity) during a positive speaking condition than CWNS (Jones, Buhr, et al., 2014) and (b) 3-year-old CWS exhibited greater sympathetic activity during a stressful picture naming task than their CWNS peers (Zengin-Bolatkale et al., 2015). These between-group differences in covert expressions of emotion (i.e., physiology) support the growing evidence of differences between young CWS and CWNS regarding overt expressions of emotion. Within-group analyses have not indicated an association between stuttering frequency and physiological indices of sympathetic activity (Choi et al., 2016); however, this association has not been investigated relative to parasympathetic activity. Thus, one purpose of the present study was to assess whether emotion regulation, indexed physiologically by respiratory sinus arrhythmia (RSA), is related to CWS's frequency of stuttering.

Respiratory Sinus Arrhythmia as an Index of Emotion Regulation

RSA is a well-substantiated measure of parasympathetic nervous system (PNS) activity on the heart mediated by the myelinated vagus (for review, see Porges, 2007). RSA is operationally defined as the amplitude of periodic fluctuations in heart rate occurring at approximately the frequency of spontaneous respiration. Porges (2001, 2007) described parasympathetic regulation of the heart—as measured by RSA—to be a useful physiological index of emotion regulation that supports behavioral processes. Such vagal (i.e., parasympathetic) influence on the heart has been likened to a "brake" (i.e., "vagal brake") that can be engaged and disengaged to promote rapid changes in cardiovascular output to meet environmental demands (Porges, Doussard-Roosevelt, Portales, & Greenspan, 1996).

According to Porges (2007), high baseline RSA acts as a brake on the heart that fosters visceral needs in the absence of external challenge and supports an individual's ability to regulate visceral state in a variety of situations. Therefore, baseline RSA has been described as an index of an individual's stress vulnerability, with higher levels supporting the ability to flexibly respond in the service of adaptive functioning. Empirical studies have indicated that higher levels of baseline RSA are associated with increased abilities to sustain and focus attention, engage in social communication, and regulate emotion (Beauchaine, Gatzke-Kopp, & Mead, 2007; Calkins, 1997; Hastings, Kahle, &

Nuselovici, 2014; Marcovitch et al., 2010; Porges, 1992; Staton, El-Sheikh, & Buckhalt, 2009).

During challenging conditions, RSA decreases as the vagal brake is released and heart rate increases, facilitating a person's ability to mobilize physiological resources to cope with environmental challenges. In this way, transitory changes in RSA serve as a component of regulatory responses during challenge and have been associated with cognitive, attentional, behavioral, emotional, and social challenges (Bar-Haim, Fox, VanMeenen, & Marshall, 2004; Gentzler, Santucci, Kovacs, & Fox, 2009; Gilissen, Koolstra, van Ijzendoorn, Bakermans-Kranenburg, & van der Veer, 2007; Heilman et al., 2008; Marcovitch et al., 2010; Weber, Van Der Molen, & Molenaar, 1994). In general, larger decreases in RSA from baseline have been associated with more adaptive responses during emotional challenges (Bar-Haim et al., 2004; Gentzler et al., 2009); however, both low and moderate decreases in RSA have been associated with better executive functioning and cognitive performance (Marcovitch et al., 2010; Utendale et al., 2014). Therefore, it is possible that the degree of decrease in RSA that would facilitate adaptive performance may be somewhat task dependent.

Inhibitory Control, Attention, and Emotion

It is not surprising that empirical evidence supports an association between RSA and components of executive functioning such as inhibitory control and attention (Becker et al., 2012; Chapman, Woltering, Lamm, & Lewis, 2010; Marcovitch et al., 2010; Staton et al., 2009; Utendale et al., 2014). This is because both RSA and aspects of executive functions are thought to interact with emotion and support emotion regulation (e.g., Gross, 2002; Gross & Thompson, 2007; Peers, Simons, & Lawrence, 2013; Rothbart, Ahadi, & Evans, 2000; White, Helfinstein, Reeb-Sutherland, Degnan, & Fox, 2009). For example, attentional biases and attention control have been found to assist in emotion regulation by facilitating scanning, monitoring, and attenuating responses to threatening stimuli (Derryberry & Reed, 2002). In schoolage children, the relation between negative affect and attention allocation to threatening stimuli appears to be mediated by effortful control, the latter involving an individual's ability to adaptively shift and focus attention, control behavior (i.e., inhibitory control), and regulate emotions (Lonigan & Vasey, 2008). Research findings indicate that children 24-36 months of age exhibit higher negative affect in association with lower effortful control (Gerardi-Caulton, 2000). Relatedly, Carlson and Wang (2007) reported that individual differences in inhibitory control were significantly correlated with preschool-age children's ability to regulate their emotions. In addition, White, McDermott, Degnan, Henderson, and Fox (2011) reported that higher levels of attention shifting and lower levels of inhibitory control were associated with lower anxiety in children with high behavioral inhibition. This brief review highlights the role that both attention regulation and inhibitory control appear to play in the regulation of emotions and temperamental reactivity.

Inhibitory Control, Attention, and Stuttering

Perhaps young CWS's apparent challenges with effortful control or emotional regulation relate to their inhibitory control (i.e., inhibiting a dominant response) to perform a subdominant response (activation control), as well as their ability to adaptively focus and/or shift attention. Empirical findings based on caregiver reports indicate that CWS, compared to CWNS, display lower attention shifting and inhibitory control (Eggers, De Nil, & Van den Bergh, 2010) and poorer attention regulation (Felsenfeld, van Beijsterveldt, & Boomsma, 2010; Karrass et al., 2006). Further, Kraft, Ambrose, and Chon (2014) reported that caregiver report of high effortful control—one aspect of which involves children's inhibitory control and ability to shift and focus attention adaptively (Rothbart, Ahadi, Hershey, & Fisher, 2001)—was related to low mother- and clinician-reported stuttering severity. Employing behavioral observation or experimental testing, it has been been reported that CWS exhibit: (a) difficulty habituating to irrelevant environmental stimuli (Schwenk et al., 2007), (b) less efficiency of the orienting subsystem of the attentional network (Eggers, De Nil, & Van den Bergh, 2012; cf. Johnson, Conture, & Walden, 2012), and (c) lower inhibitory control during a Go/NoGo task (Eggers, De Nil, & Van den Bergh, 2013). Clark, Conture, Walden, and Lambert (2015) also reported that for CWS, but not CWNS, lower attentional distractibility was associated with more speech-language dissociations (i.e., imbalances among subcomponents of speech-language planning and production). Thus, there is mounting evidence that inhibitory control and attention differentiate CWS from CWNS.

Conture and Walden (2012) proposed one possible mechanism by which emotion and cognitive processes may contribute to instances of stuttering. Specifically, emotion occurring concurrently with speech processes may unduly draw on finite cognitive resources necessary to support the initiation and/or maintenance of fluent speech-language planning and production. However, empirical study of such speculation has been scant. One study reported that behavioral observations of emotion just prior to and during initiation of speech increased the likelihood of stuttering in subsequent utterances (Jones, Conture, & Walden, 2014). These authors suggested that emotion immediately prior to and/or at the time of speech initiation—a period theoretically associated with many speech-language processes (for review, see Levelt, Roelofs, & Meyer, 1999)—may divert and/or disrupt cognitive resources necessary for rapid, efficient speech-language planning and thus contribute to stuttering.

The Current Study: Emotion Regulation, **Executive Functions, and Childhood Stuttering**

The current study investigated whether RSA was associated with stuttering frequency for young CWS and CWNS. In addition, we also investigated whether these RSA-stuttering associations were moderated by executive

functions. In order to address these issues, the present study employed six conditions: (a) viewing a neutral (baseline) video clip, (b) viewing two emotionally arousing video clips (positive and negative), and (c) three narrative speaking tasks that immediately followed the viewing of each video clip (neutral, positive, negative).

Table 1 provides an overview of the three RSA-related measures: (a) baseline RSA (i.e., RSA measured during environmental situation involving no external challenge), (b) RSA change during video viewing conditions (i.e., change in RSA from baseline to video clip viewing), and (c) RSA change during speaking conditions (i.e., change in RSA from baseline to speaking). This table also explains that values of baseline RSA are discussed as "high"/"low" and that values of RSA change are discussed as "increased"/"decreased." The present study also measured frequency of stuttered disfluencies during the three narrative speaking conditions, as well as executive functions, the latter indexed by caregiverreported individual differences in inhibitory control and attentional focusing.

Based on the above review and the authors' own empirical study of emotion and childhood stuttering (Choi et al., 2016; Jones, Buhr, et al., 2014), we speculated that executive functions (i.e., composite of inhibitory control and attentional focusing) may serve as a potential link between psychophysiological emotion processes and disruptions in young CWS's speech-language fluency (Conture & Walden, 2012). To determine the support for such speculation, we explored four related but different questions and their four associated testable hypotheses.

The first question involved whether there is an association between young CWS and CWNS's baseline RSA and stuttering in subsequent speaking conditions. We hypothesized that for CWS, when compared to their CWNS peers, lower baseline RSA would be significantly associated with increased stuttering frequency during subsequent speaking conditions (especially when such speaking followed viewing of emotionally arousing video clips).

The second question involved whether there is an association between young CWS and CWNS's RSA change during video viewing and stuttering during subsequent speaking conditions. We hypothesized that for CWS, compared to their CWNS peers, greater decreases in RSA during video viewing of emotionally arousing video clips (positive and negative), would be significantly associated with increased stuttering frequency during subsequent speaking conditions.

The third question involved whether there is an association between young CWS and CWNS's RSA change and stuttering during speaking conditions. We hypothesized that for CWS, when compared to their CWNS peers, greater decreases in RSA during speaking would be significantly associated with increased stuttering frequency, particularly following emotionally arousing (compared to neutral) video clips.

The fourth and final question involved whether there is an association between young CWS and CWNS's executive functions and RSA-stuttering relation. We hypothesized the stuttering of CWS with lower executive functions would be impacted by physiological state (i.e., change in RSA)

Table 1. Description of the present study's (a) three dependent measures of respiratory sinus arrhythmia (RSA), (b) what the RSA measure indexed, (c) associated physiological action, (d) what they are operationalized by, and (e) terminology and hypothetical examples.

(a) Dependent measure of RSA	(b) Index	(c) Physiological action	(d) Operationalized by	(e) Terminology used in present study	(f) Hypothetical example of finding
Baseline RSA	Stress vulnerability; trait-like aspect of emotion regulation	Tonic parasympathetic activity on the heart (i.e., while neither speaking during narrative task nor viewing emotionally arousing video clips)	Raw RSA values measured during baseline (neutral) condition (e.g., fish tank screen saver in the current study)	Discussed as level of RSA (e.g., "high"/"low")	Lower baseline RSA was not significantly associated with increased stuttering frequency during narrative/speaking task.
RSA change during video viewing	Regulatory responses during video clip viewing of positive and negative emotion stimuli	Transitory shifts in parasympathetic activity during emotional challenge (i.e., while viewing emotionally arousing video clips)	Residualized RSA change score (RSA during video viewing – Baseline RSA)	Discussed as directionality of RSA change (e.g., "increase"/ "decrease")	Greater decrease of RSA during video viewing was significantly associated with increased stuttering during narrative/ speaking task.
RSA change during speaking	Regulatory responses during speaking following baseline and emotional video clip viewing tasks	Transitory shifts in parasympathetic activity during speaking (i.e., while speaking during narrative task following baseline and emotionally arousing video clips)	Residualized RSA change score (RSA during speaking task – Baseline RSA)	Discussed as directionality of RSA change (e.g., "increase"/ "decrease")	Greater decrease of RSA during speaking was significantly

more than those with higher executive functions, and this association would significantly differ from that of CWNS.

Method

Participants

Participants¹ were 36 young CWS (28 boys, 8 girls) and 36 young CWNS (19 boys, 17 girls) 36-71 months of age. Chronological age for CWS (M = 47.31, SD = 8.72) did not significantly differ from CWNS (M = 49.69, SD =8.62). Participants' race and socioeconomic status (SES) were obtained via caregiver interviews. There was no significant between-group difference in SES. CWS were 28 Caucasians, six African Americans, one Asian, and one child with more than one reported race. CWNS were 32 Caucasians, one Native American/Alaskan, and three children with more than one reported race.

From an initial pool of 88 participants (44 CWS, 42 CWNS, two unclassifiable), two children were excluded because their talker group classification (CWS/CWNS) was undeterminable. Twelve participants (six CWS and six CWNS) were excluded because it was not possible to acquire their RSA due to noncompliance and/or excessive data artifact. Two participants' (CWS) data were not included because they were deemed to be outliers, as described below in the Pre-Analysis Data Preparation section. There were no significant differences between excluded participants and those who were included in the study on standardized speech-language tests or SES. This resulted in a total of 36 CWS and 36 CWNS contributing to the final data corpus.

All participants were paid volunteers naïve to the purposes and methods of the study. Most participants were recruited through a longitudinal study of emotional and linguistic contributions to childhood stuttering conducted by Vanderbilt University's Developmental Stuttering Project (e.g., Choi et al., 2016; Clark et al., 2015; Jones, Buhr, et al., 2014; Ntourou et al., 2013). Participants were referred by their caregivers who were informed via (a) a free, widely read parent-oriented magazine, (b) local health care providers, or (c) self/professional referrals to the Vanderbilt Bill Wilkerson Hearing and Speech Center.

Vanderbilt University's Institutional Review Board (IRB) approved the protocol, and informed consent by parents and assent by children were obtained.

Classification and Inclusion Criteria

On a separate day, and prior to experimental testing, children participated in a diagnostic visit. At this visit, children engaged in a 300-word conversational speech sample

with an examiner and were administered standardized speech-language assessments.

Children classified as CWS exhibited three or more stuttered disfluencies (i.e., sound syllable repetitions, monosyllabic whole-word repetitions, and sound prolongations) per 100 words of conversational speech (Conture, 2001; Tumanova, Conture, Lambert, & Walden, 2014; Yaruss, 1997, 1998) and received an overall score of 11 or higher on the Stuttering Severity Instrument for Children and Adults-Third Edition (SSI-3; Riley, 1994). Children classified as CWNS exhibited two or fewer stuttered disfluenices per 100 words of conversational speech and received an overall score of 10 or below on the SSI-3. No CWS was reported or known to have received or was receiving treatment for stuttering.

All participants scored at or above the 16th percentile on standardized speech and language tests, including the Goldman Fristoe Test of Articulation-Second Edition (GFTA-2; Goldman & Fristoe, 2000), Peabody Picture Vocabulary Test-Fourth Edition (PPVT-4; Dunn & Dunn, 2007), the Expressive Vocabulary Test–Second Edition (EVT-2; Williams, 2007), and the Test of Early Language Development-Third Edition (TELD-3; Hresko, Reid, & Hammill, 1999). SES was determined by scoring parent education and occupation levels on the Hollingshead Index (Hollingshead, 1975).

One-way ANOVA indicated that the CWS (M = 115.61, SD = 11.53) scored significantly lower on the EVT-2² than CWNS (M = 122.81, SD = 10.70), F(1, 70) = 7.531, p = .008, $\eta^2 = 0.097$. There were no significant differences between the two talker groups on any other standardized measure of speech-language abilities.

Procedure

As described in a similar study employing different dependent variables and testing different hypotheses (e.g., Jones, Buhr, et al., 2014, p. 18), participants were seated in a child-sized "jeep" in a car safety seat directly in front of a computer monitor. After allowing a few minutes to acclimate, the participant's skin was cleaned with alcohol to optimize electrical conductance of electrodes. Hypoallergenic electrodes were applied to the skin surface at the suprasternal notch of the rib cage and at the 12th rib laterally on the left for acquisition of heart rate. A lapel microphone attached to the participant's clothing acquired the audio signal.

For the preexperimental neutral (baseline) condition, participants viewed an animated screensaver of a fish tank for 4 minutes. The screensaver contained minimal action and was assumed to be suitable to establish baseline RSA. The 4-minute duration of neutral stimuli (i.e., fish tank screensaver) is consistent with previous studies that have

¹Some participants' data (20 CWS and 21 CWNS) included in the present study were also in the Jones, Buhr, et al. (2014) study, with this previous empirical study involving different dependent variables (e.g., between-group comparisons in level of RSA) and hypotheses (none overlapping) from the present study.

²The EVT-2 was seemingly of limited relation to the association between the variables under study (e.g., RSA and stuttering). Nevertheless, all analytical models testing the a priori hypotheses were conducted with and without EVT-2 as a covariate, and statistical significance remained the same. Thus, in Results, we present findings of analytical models without EVT-2 as a covariate.

measured baseline RSA (e.g., Bar-Haim et al., 2004; Gentzler et al., 2009).

Figure 1 provides a visual representation of the experimental sequence of the aforementioned six conditions. In brief, after the preexperimental neutral (baseline) condition, participants listened to and viewed one negative and one positive 4-minute audio/video clip from one of five movies appropriate for children, including Happy Feet, The Lion King, The Little Mermaid, The Wizard of Oz, and The Princess and the Frog. Each video clip was intended to elicit negative or positive emotion. Order of presentation of video clips was counterbalanced across children. After the neutral (baseline) and each of the two emotional video clips, participants were asked to tell a story (narrative task) about one of three textless storybooks (unrelated to the audio/video clips) about a boy, a dog, and a frog by the author Mercer Mayer (e.g., Frog Where Are You?; Mayer, 1969). To control for potential differential influences on participants' speech (dis)fluency, the order of presentation of the various storybooks was counterbalanced across children.

The effectiveness of the audio/video clips in eliciting the expected positive, negative, and neutral emotions was validated by research personnel trained in coding of emotional expression in young children from videotape (one PhD student and one master's-level research assistant). Blind to experimental condition, these research personnel viewed video recordings of eight preschool-age CWS and eight of their CWNS peers during each of four experimental conditions (neutral, negative, positive) and made a judgment as to what the children were viewing. They accurately identified the valence of the video (neutral, positive, negative) for 79% of conditions. Thus, the experimental conditions had differential effects specific to each emotion condition. These behavioral observations assessed emotional responses rather than self-reports because children have difficulty reliably reporting their emotional states at this age.

Measures

Executive Functions

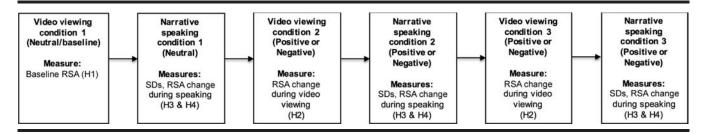
The 195-item version of the Children's Behavior Questionnaire (CBQ; Rothbart et al., 2001) is a commonly used questionnaire to assess parent report of temperament in children 3-7 years of age, and it has been used in empirical studies of the temperament of young CWS (Choi et al., 2016; Eggers et al., 2010; Walden et al., 2012). Parents rated whether each of the 195 items was true of their child (rated from 1 = extremely untrue to <math>7 = extremely true). In the present study, the inhibitory control (13 items) and the attentional focusing (nine items) subscales were significantly correlated (r = .68, p < .0001) and were combined to create a composite score used to index executive functions. Cronbach's alpha for the executive functions composite score was .87.

Respiratory Sinus Arrhythmia

As mentioned above, RSA is operationally defined as the amplitude of periodic fluctuations in heart rate occurring at approximately the frequency of spontaneous respiration. To obtain RSA, as described in Jones, Buhr, et al. (2014), an electrocardiogram (ECG) was acquired for each participant using the Biopac MP150 system (Biopac Systems, Inc., Goleta, CA) and digitized at 1250 Hz. The raw ECG was band-pass filtered to remove high frequency noise and low frequency drift (high pass cutoff: 0.5 Hz; low pass cutoff: 35 Hz). Biopac software processed the ECG signal by detecting the peak of the R-wave and timing the sequential interbeat intervals (IBI) in milliseconds. IBI time series were produced from segments corresponding to each of the three video viewing conditions (e.g., neutral, positive, negative) as well as three narrative speaking conditions.

The IBI time series were processed by CardioEdit software (CardioEdit, 2007) to correct artifacts due to ventricular arrhythmias and movement by adding, dividing, or averaging consecutive points to be consistent with surrounding points. The RSA or sinus atrial node rhythm can be distorted when ventricular rhythms are present in the IBI time series (measured in milliseconds). No more than 5% of the total RSA data for any condition were corrected using CardioEdit. CardioBatch software (CardioBatch, 2007) was used to derive measures of RSA from corrected IBI files. Respiratory sinus arrhythmia was calculated using Porges' method (Porges, 1985; Porges & Bohrer,

Figure 1. This figure illustrates the experimental sequence of the six conditions (i.e., three video viewing conditions [baseline/neutral, negative, and positive] and three narrative speaking conditions [neutral, negative, and positive] and their associated repeated measures—stuttered disfluencies [SDs] and respiratory sinus arrhythmia [RSA]). The figure depicts the study's dependent measures for one baseline/neutral video viewing (i.e., baseline RSA), two video viewings of emotionally arousing (positive and negative) video clips (i.e., RSA change during video viewing), and three speaking tasks (i.e., RSA change during speaking and SDs). As noted in the prose, the order of the negative video viewingnegative speaking and positive video viewing-positive speaking conditions was counterbalanced across all participants. These dependent measures were used to test the study's four hypotheses (H1, H2, H3, H4).



1990), which time samples the IBI time series into 250-ms samples and uses a 21-point polynomial to detrend periodicities and trends in the IBI series slower than RSA (e.g., ultradians reflecting basic metabolic processes).

A band-pass filter was applied to the time-sampled IBI series (i.e., 250-ms intervals) to extract variance at the frequency of respiration for young children (0.24–1.04 Hz). RSA was expressed as the natural log of this variance: ln (ms)² (for further details, see Lewis, Furman, McCool, & Porges, 2012). Values of RSA were derived for sequential 30-s epochs within each 4-min condition (i.e., associated with neutral and emotionally arousing video clips, and narrative speaking tasks). The average of the 30-s epochs within each 4-min epoch was used as the metric for RSA in the inferential analyses of data. In each statistical model using RSA change, change scores adjusted for the baseline value were calculated with regression analysis to account for the initial value (law of initial values; Wilder, 1962) and is a common procedure used in the study of RSA (e.g., Heilman et al., 2008; see Table 1).

Speech Disfluencies

The first author and four trained speech-language pathologists (SLPs) counted the number of stuttered disfluencies during each of the three speaking conditions, that is, during each narrative speaking task following the baseline, positive, and negative video viewing conditions. Stuttered disfluencies included: (a) sound/syllable repetitions (e.g., "b-boy," "fr-frog"); (b) monosyllabic whole-word repetitions (e.g., "I-I-I," "he-he-he"); and (c) sound prolongations (e.g., "mmmm-me," "ssssss-sand"). Although the CWNS produced a sufficient number of stuttered disfluencies during the narrative for statistical analysis, none had a frequency of stuttered disfluencies that rose to the threshold for a diagnosis of stuttering (the conversational sample mentioned in the Classification and Inclusion Criteria section was used for talker group classification).

In the present study, and as shown elsewhere (Tumanova et al., 2014), stuttered speech disfluencies were distributed as a negative binomial. Thus, a transformation (stuttered disfluencies + 1) and a statistical approach appropriate for analyzing such distributions was employed (described below).

Measurement Reliability for RSA and Stuttered Disfluencies

Twenty-four percent of the total final data (data from 17 of 72 total participants) were randomly selected to determine interrater reliability between the first author and trained researchers (graduate-level or above) for RSA. For RSA, values of RSA for pairs of raters were within 0.10 ln (ms)² for all conditions, with correlations of RSA values between pairs of raters exceeding .99.

For stuttered disfluencies, intraclass correlation coefficients (ICC; McGraw & Wong, 1996; Shrout & Fleiss, 1979) using the absolute agreement criterion were computed to assess interjudgment reliability for the measurement of

stuttered disfluencies. Interjudge agreement reflected agreement between the first author and four trained coders independently judging stuttered disfluencies for 29 participants from video-recorded speech samples. The 95% confidence intervals (CI) ranged from .91-.97, with ICC measures of .95, p < .001, for identification of stuttered disfluencies.

Preanalysis Data Preparation

The frequency of stuttered disfluencies during the three speaking conditions following the neutral, positive, and negative video viewing conditions was examined for outliers and potentially influential values. Similar to studies of reaction time in young CWS and CWNS (e.g., Johnson et al., 2012), we excluded data from participants who exhibited a frequency of stuttered disfluencies greater than three SDs above the mean. This is a recommended criterion by Osborne and Overbay (2004).

This criterion for outliers was employed to increase the likelihood that the data are representative of the population as a whole, while also decreasing the likelihood of an error of inference (Osborne & Overbay, 2004), that is, the situation in which a few extreme cases might drive a significant result. As mentioned in the Participants section, this criterion led to the exclusion of two CWS, which resulted in a final data corpus of 36 CWS and 36 CWNS for the present data analysis.

Data Analysis

Inspection of histograms and the Shapiro-Wilk W test (Razali & Wah, 2011) assessed the normality of the distribution of speech disfluencies, the dependent variable for each hypothesis. As mentioned above, and similar to Tumanova et al. (2014), stuttered disfluencies were not normally distributed, displaying high positive skew (skewness = 1.822, W = .765, df = 216, p < .0001). To account for nonnormality, generalized linear models with generalized estimating equations (GEE) were used (Zeger & Liang, 1986; Zeger, Liang, & Albert, 1988) with negative binomial distribution and log-link function. "Negative binomial" refers to a skewed distribution similar to Poisson but with an overdispersion parameter to measure the long right-hand tail. Negative binomial is the preferred specification for statistically assessing counts of events with this type of distribution (Gardner, Mulvey, & Shaw, 1995). This form of negative binomial regression in exponential form is nonlinear (Aiken, Mistler, Coxe, & West, 2015) and can be thought of as multiplicative (Atkins, Baldwin, Zheng, Gallop, & Neighbors, 2013). Therefore, exponentiated coefficients "provide a multiplying factor for each unit of change in the covariate" (Atkins et al., 2013, p. 6), whereas linear models are additive. Accordingly, plots of predicted counts from the negative binomial regressions—as shown in Results-are curvilinear.

Statistical Modeling

The GEE models, used in the present study, control for correlated errors occurring with repeated measures

(Nich & Carroll, 1997), and they were run using the GENMOD procedure of SAS version 9.4 for Windows (SAS Institute, Cary, NC). The four a priori hypotheses tested differences in young CWS and CWNS's association between baseline RSA and stuttering frequency (Hypothesis 1), RSA change during video viewing and stuttering frequency (Hypothesis 2), RSA change during speaking and stuttering frequency (Hypothesis 3), as well as the emotion condition (Hypotheses 1, 2, and 3), and a potential moderator, executive functions (Hypothesis 4). Moderation analysis statistically specifies the conditions under which two variables (e.g., RSA change during speaking and stuttering) are associated with one another as a function of a third variable (e.g., executive functions) (Kraemer, Wilson, Fairburn, & Agras, 2002).

Thus, moderation analyses assessed the nature of the significant interaction effects of interest to this study's hypotheses (e.g., interaction of RSA Change During Speaking × Executive Functions × Group). For significant interactions relating to moderation analyses, simple regressions of the predictor variable (RSA change during speaking) on the dependent variable (stuttering frequency) were calculated and tested for significance at three different levels (one SD below mean, at the mean, and one SD above mean) of the moderator variable (e.g., executive functions) as described by Aiken and West (1991) and Holmbeck (2002). To avoid issues with multicollinearity, all continuous predictor variables (e.g., X) and covariates (e.g., Y) were mean centered (Aiken & West, 1991). A measure of effect size (R^2) was estimated for each model by calculating the Pearson's rho between the observed and modeled (predicted) stuttered disfluencies (Singer & Willett, 2003). R^2 is a measure of how well the proportion of total variance of stuttered disfluencies is accounted for by the model, and values of .01/.09/.25 may be considered small/medium/large effect sizes, respectively (Cohen, 1988).

Statistical Models for Each of the Study's Four a Priori Hypotheses

Hypothesis 1: For CWS, Compared to CWNS, Lower Baseline RSA Would Be Significantly Associated With Increased Stuttering During Subsequent **Speaking Conditions**

To address the first hypothesis, one model was constructed to assess between-group differences in the relation between young CWS and CWNS's baseline RSA (assessed while participants watched a neutral movie clip) and their frequency of stuttered disfluencies during the subsequent three speaking conditions. The between-group model included baseline RSA, group, condition (neutral, positive, negative), and of particular interest the interaction of Baseline RSA \times Group \times Condition (and all other two-way interactions: Baseline RSA × Condition, Baseline RSA × Group, Group × Condition) to predict the dependent variable of stuttered disfluencies. This model allowed us to determine between-group differences in the relation between stuttering frequency and baseline RSA.

Hypothesis 2: For CWS, Compared to CWNS, Greater Decreases in RSA During Video Viewing Conditions Would **Be Significantly Associated With Increased Stuttering**

To address the second hypothesis, one model was constructed to assess between-group differences in the relation between young CWS and CWNS's RSA change during the two emotionally arousing video clip viewing conditions (positive, negative) and the frequency of their stuttered disfluencies during subsequent speaking conditions. The between-group model included RSA change, group, condition (positive, negative), and of particular interest the interaction of RSA Change × Group × Condition (and all other two-way interactions: RSA Change × Condition, RSA Change × Group, Group × Condition) to predict the dependent variable of stuttered disfluencies. This model allowed us to determine between-group differences in the relation between frequency of stuttered disfluencies and RSA change during positive and negative video viewing.

Hypothesis 3: For CWS, Compared to CWNS, Greater Decreases in RSA During Speaking Would Be Significantly **Associated With Increased Stuttering**

To address the third hypothesis, one model was constructed to assess between-group differences in the relation between young CWS and CWNS's RSA change during each speaking condition and the frequency of their stuttered disfluencies. Similar to the second hypothesis, the between-group model included RSA change, group, condition (neutral, positive and negative), and the interaction of RSA Change × Group × Condition (and all other twoway interactions: RSA Change × Condition, RSA Change × Group, Group × Condition) to predict the dependent variable of stuttered disfluencies. This model allowed us to determine between-group differences in the relation between frequency of stuttered disfluencies and RSA change during speaking.

Hypothesis 4: The Stuttering of CWS With Lower Executive Functions Would Be Impacted by Physiological State (i.e., Change in RSA) More Than Those With Higher Executive Functions, and This Association Would Significantly Differ From That of CWNS

To address the fourth hypothesis, one model was constructed to examine whether executive functions moderated the relation between young CWS and CWNS's RSA change during the three narrative speech conditions and the frequency of their stuttered disfluencies. We developed a between-group model with RSA change, executive functions, group, and the interaction of RSA Change × Executive Functions × Group (as well as all other two-way interactions: RSA Change × Group, RSA Change × Executive Functions, Executive Functions × Group) to predict the dependent variable of stuttered disfluencies. This model allowed us to determine how the frequency of stuttered disfluencies of CWS and CWNS differ as a function of the relation between executive functions and RSA change.

Fixed Factors and Continuous Covariates

In general, four covariates were included in all statistical models: (a) gender, (b) chronological age, (c) respiratory frequency, and (d) total number of words spoken. Specifically, one fixed factor, gender, was covaried in each model. An estimate of respiration rate was derived from the beat-to-beat heart rate pattern using a custom program, RespFreqFromRSA (Lewis, 2010) and entered in each model as a covariate. The frequency of RSA maps onto the frequency of respiration (correlation above .99), when respiration is monitored via changes in chest circumference (see Denver, Reed, & Porges, 2007). Since vocalizations are dependent on breathing, respiratory frequency may also be associated with stuttered disfluencies. Age in months was entered as a continuous covariate (known to be related to RSA; El-Sheikh, 2005). Given that the total number of words produced by each child varied during the narrative speaking tasks, total number of words was also covaried to account for the impact of total number of words spoken on number of stuttered disfluencies.

Results

Descriptive Analyses

Speech Fluency

Between-group differences in conversational speech fluency during the diagnostic visit (used for group classification) were evaluated with ANOVA. Consistent with talker group classification criteria, there was a significant difference in stuttered disfluencies per 100 words between CWS (M = 8.3, SD = 4.4) and CWNS (M = 1.1, SD = 0.7), $F(1, 70) = 93.9, p < .001, \eta^2 = .58$, and a difference in stuttering severity (i.e., SSI-3 scores) between CWS (M = 18.4, SD = 5.8) and CWNS (M = 6.3, SD = 1.9), F(1, 70) =142.8, p < .001, $\eta^2 = .67$. In addition, there was a significant difference between CWS (M = 13.4, SD = 5.1) and CWNS $(M = 4.5, SD = 2.7), F(1, 70) = 83.7, p < .001, \eta^2 = .55 \text{ in}$ total disfluencies per 100 words, and between CWS (M = 5.0, SD = 3.1) and CWNS (M = 3.4, SD = 2.5), F(1, 70) = 5.7, p = .02, $\eta^2 = .08$ in nonstuttered disfluencies per 100 words.

Respiration

A linear mixed-effects model (LMM; Pinheiro & Bates, 2013) examined whether respiration rate differed between CWS and CWNS, particularly during the emotionally arousing video clips (nonspeaking) versus narrative speaking tasks (speaking). First, for all participants (i.e., CWS + CWNS), there was no significant difference in respiration rate between narrative speaking conditions (estimated marginal mean, EMM = .382, standard error, SE = .003) and viewing of emotionally arousing video conditions (EMM = .390, SE = .005), F(1, 66.00) = 2.38, p = .128.Second, a between-group (i.e., CWS vs. CWNS) comparison indicated no significant difference in respiration rate during all narrative speaking conditions and viewing of emotionally arousing video conditions, CWS (EMM = .389, SE = .005), CWNS (EMM = .383, SE = .005),

F(1, 66.48) = .59, p = .444. Last, there was no significant interaction of group by speech condition (i.e., speaking vs. nonspeaking condition) for respiration rate, $F(1, \frac{1}{2})$ 66.00) = 1.89, p = .174. These between-group as well as between speaking and nonspeaking similarities in respiratory rate suggest that respiration cannot readily account for subsequent observed between-group differences in the relation between RSA (baseline RSA, RSA change) and stuttered disfluencies.

Inferential Analyses Regarding a Priori Hypotheses

Results pertaining to the four a priori hypotheses are presented below. Table 2 provides descriptive statistics for speech disfluencies, measures of RSA, and the composite measure of executive functions.

Hypothesis 1: For CWS, Compared to CWNS, Lower **Baseline RSA Would Be Significantly Associated** With Increased Stuttering During Subsequent **Speaking Conditions**

Contrary to predictions, there was no between-group difference (i.e., CWS vs. CWNS) in the relation between stuttered disfluencies and baseline RSA (p = .596; Baseline RSA × Group), or in the relation between stuttered disfluencies and baseline RSA as a function of emotionally arousing video viewing conditions (p = .173; Baseline RSA \times Group × Condition). Baseline RSA, on its own, was not predictive of stuttered disfluencies (p = .204) in speaking conditions for either CWS or CWNS. These results did not support Hypothesis 1.

Hypothesis 2: For CWS, Compared to CWNS, Greater Decreases in RSA During Video Viewing Conditions Would **Be Significantly Associated With Increased Stuttering**

There was no statistically significant between-group difference (i.e., CWS vs. CWNS) in the relation between stuttered disfluencies and RSA change during the negative and positive emotionally arousing video viewing conditions $(p = .077; RSA Change \times Group \times Condition)$. However, the size of the p value (i.e., p = .077) warrants further exploration, since the absence of an effect might be an issue of insufficient power. Parameter estimates for RSA change were statistically significant only for CWS during the positive video clip (CWS: $\beta = -.46$, 95% CI = -.69 to -.24, p = < .0001).

As predicted in Hypothesis 2, there was a negative association between RSA change during the emotionally arousing video viewing conditions and stuttered disfluencies for both groups, Wald χ^2 (1) = 7.83, p = .005, $R^2 = .34$ (CWS: $\beta = -.24$, 95% CI = -.51 to .04, p = .091; CWNS: $\beta = -.22$, 95% CI = -.45 to .01, p = .065). Greater decrease of RSA during the emotionally arousing video viewing conditions was related to more stuttered disfluencies during narratives that followed each condition, a pattern that did not significantly differ between the two talker groups. These results partially supported Hypothesis 2.

Table 2. Descriptive statistics—means (M) and standard deviations (SD)—associated with the present study's six conditions (i.e, three video viewing and three speaking).

	RSA	RSA change	Stuttered disfluencies	Executive Functions M (SD)
Condition	M (SD)	M (SD)	M (SD)	
Children who stutter ($n = 36$)				
Neutral video viewing (baseline)	6.19 (1.17)	_	_	4.36 (.72)
Neutral narrative speaking	5.72 (1.14)	47 (0.48)	6.54 (5.13)	_ ` ´
Positive video viewing	6.23 (1.43)	.04 (0.65)	_` ´	_
Positive narrative speaking	5.36 (1.06)	82 (0.53)	6.06 (5.74)	_
Negative video viewing	6.18 (1.43)	01 (0.61)	<u>—</u> `	_
Negative narrative speaking	5.46 (1.13)	72 (0.65)	6.31 (5.95)	_
Children who do not stutter ($n = 36$)				
Neutral video viewing (baseline)	6.61 (1.10)	_	_	4.69 (.81)
Neutral narrative speaking	6.06 (0.92)	54 (0.41)	1.87 (1.32)	_ ` `
Positive video viewing	6.63 (0.98)	.02 (0.70)	<u>—</u> `	_
Positive narrative speaking	5.80 (0.86)	81 (0.58)	1.68 (1.74)	_
Negative video viewing	6.43 (1.06)	18 (0.71)	<u>—</u> `	_
Negative narrative speaking	5.81 (0.80)	80 (0.59)	1.64 (1.52)	_

Note. First, the table shows the M and SD for RSA (respiratory sinus arrhythmia) values for each of the six conditions. Second, the table shows the M and SD for RSA change from baseline during emotionally arousing video viewing conditions (positive and negative) and narrative speaking conditions, the latter which immediately followed video viewing conditions (neutral, positive, negative). Third, the table shows the M and SD for stuttered disfluencies per 100 words for each of the three speaking conditions. Fourth, and finally, the table shows the M and SD for executive functions, that is, a composite score of the inhibitory control and attentional focusing from the Children's Behavior Questionnaire (Rothbart, Ahadi, Hershey, & Fisher, 2001) for young children who stutter and children who do not stutter. It should also be noted, for the purposes of general description, that this table presents raw means and standard deviations, uncorrected for covariates.

Hypothesis 3: For CWS, Compared to CWNS, Greater Decreases in RSA During Speaking Would Be Significantly **Associated With Increased Stuttering**

Similar to the findings for Hypothesis 2, there was no between-group difference in the relation between stuttered disfluencies and RSA change during the speaking conditions as an effect of condition (p = .850; RSA × Group \times Condition).

As with findings pertaining to video viewing, there was a negative association between RSA change during speaking and stuttered disfluencies for both talker groups, Wald χ^2 (1) = 4.98, p = .026, $R^2 = .39$ (CWS: $\beta = -.41$, 95% CI = -.60 to -.22, p < .0001; CWNS: $\beta = -.07$, 95% CI = -.55 to .41, p = .76). Greater decrease of RSA during speaking was related to more stuttered disfluencies, a pattern that held in both talker groups. These results partially supported Hypothesis 3.

Hypothesis 4: The Stuttering of CWS With Lower Executive Functions Would Be Impacted by Physiological State (i.e., Change in RSA) More Than Those With Higher **Executive Functions, and This Association Would** Significantly Differ From That of CWNS

Consistent with Hypothesis 4, the three-way interaction of RSA Change × Executive Functions × Group significantly predicted stuttered disfluencies, Wald $\chi^2(1)$ = 6.69, p = .009, $R^2 = .39$ (CWS: $\beta = .59$, 95% CI = .26 to .92, p < .001, CWNS: $\beta = -.43$, 95% CI = -.84 to -.02, p = .042).

First, to probe this significant three-way interaction, we conducted within-group analyses for each talker group (CWS and CWNS). This involved calculation of simple

regressions of stuttering frequency on RSA change during speaking conditions at three levels (one SD below mean, at the mean, and one SD above mean) of the moderator variable (executive functions) and determined the significance for each of the resulting six regressions (three for CWS and three for CWNS). Second, we conducted between-group (CWS vs. CWNS) comparisons of the simple regressions at each level of the moderator variable.

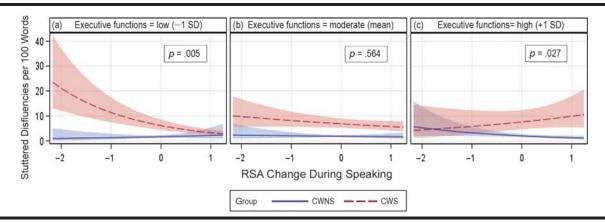
Within-group analyses. For CWS, results indicated that the simple regressions of RSA change were statistically significant at one SD below the mean ($\beta = -.65$, SE = .09, p < .0001) of executive functions, whereas they were not significant at the mean (p = .071) or one SD above the mean (p = .218).

Conversely, for CWNS, results indicated that the simple regressions of RSA change were neither significant at 1 SD below the mean (p = .376) nor at the mean (p = .810), but were significant at 1 SD above the mean ($\beta = -.38$, SE = .19, p = .045) of executive functions.

Between-group analyses. Between-group comparisons of these simple regressions indicated significant differences between CWS and CWNS at 1 SD below the mean $(\beta = .93, SE = .33, p = .005)$ and 1 SD above the mean $(\beta = -.66, SE = .30, p = .027)$ of executive functions, but not at the mean (p = .564).

Figures 2a–2c illustrate predicted counts of stuttered disfluencies across the range of RSA change for three conditional levels (low/moderate/high) of executive functions for CWS and CWNS. The predicted counts from the negative binomial regression are curvilinear (Aiken et al., 2015; Atkins et al., 2013). The degree or extent of the curve for each of these simple regressions (e.g., association between

Figure 2. Predicted counts (simple curvilinear regression lines) of stuttering frequency across the range of respiratory sinus arrhythmia (RSA) change during speaking at three conditional levels of the moderator variable (i.e., executive functions) (plots a–c) for young children who do not stutter (CWNS)—blue solid line—and children who stutter (CWS)—red dashed line. For the moderator variable, "low," "moderate," and "high" represents participants (both CWS and CWNS) who scored 1 *SD* below mean, mean, and 1 *SD* above mean for executive functions, respectively. Light blue (CWNS) and red (CWS) shaded areas represent 95% confidence limits. The *p* values reported in each plot represent between-group (CWS vs. CWNS) comparisons for the two simple regressions lines within each plot.



stuttering frequency and RSA change for CWS at 1 *SD* below the mean of executive functions) varies as a function of the nature of the association between stuttering frequency and changes in RSA (i.e., decreases/increases).

In Figure 2a (illustrating findings for 1 SD below the mean for executive functions), the simple regression depicts a steep line for CWS with low levels of executive functions, indicating that greater decrease of RSA significantly predicted higher stuttering frequency. However, the simple regression depicts a flatter line for CWNS with low levels of executive functions, indicating that decreases in RSA did not significantly predict higher stuttering frequency.

In Figure 2b (illustrating findings at the mean for executive functions), the simple regressions depict relatively flat lines for both CWS and CWNS, indicating that for both CWS and CWNS with moderate levels of executive functions, decrease of RSA did not significantly predict higher stuttering frequency.

Last, for Figure 2c (illustrating findings for 1 SD above the mean for executive functions), the simple regression depicts a significant line for CWNS with high levels of executive functions, indicating that greater decrease of RSA significantly predicted higher stuttering frequency. However, for CWS with higher levels of executive functions, decrease of RSA did not significantly predict higher stuttering frequency.

These results supported Hypothesis 4.

Summary of Main Findings

The present study's main findings indicated that for both groups, RSA change during the emotionally arousing video clips and narrative speaking tasks was significantly associated with the frequency of stuttered disfluencies. In addition, CWS with lower executive functions exhibited a significant association between RSA change and stuttering, whereas CWNS with higher executive functions exhibited a significant association between RSA change and stuttering. Last, between-group and condition differences in respiratory rate cannot readily account for the above findings given that there were neither significant differences in respiration rate between the two groups (i.e., CWS vs. CWNS) nor conditions (i.e., speech vs. nonspeech); likewise, respiration rate was not associated with stuttering frequency.

Discussion

A Priori Hypotheses

Hypothesis 1: For CWS, Compared to CWNS, Lower Baseline RSA Would Be Significantly Associated With Increased Stuttering During Subsequent Speaking Conditions

For both young CWS and their CWNS peers, there was no significant association between baseline RSA and stuttering frequency during the speaking conditions. This finding was taken to suggest that an individual's stress vulnerability, as indexed by baseline RSA, was not associated with stuttering frequency during speaking that occurred in close temporal proximity.

Hypothesis 2: For CWS, Compared to CWNS, Greater Decreases in RSA During Video Viewing Conditions Would Be Significantly Associated With Increased Stuttering

For both young CWS and their CWNS peers, there was a negative association between stuttered disfluencies during the speaking conditions and RSA change during emotionally arousing video viewing conditions. These findings indicate that greater decreases in RSA, a physiological index of emotion regulation, is associated with greater frequency of stuttered disfluencies for both CWS and CWNS.

Our initial predictions were supported by the finding that young CWS's stuttering frequency is related to a physiological index of emotion regulation. This finding is also consistent with empirical findings that young CWS's stuttering frequency is related to behavioral indices of emotion regulation (Arnold et al., 2011; Jones, Conture, et al., 2014; Kraft et al., 2014; Walden et al., 2012). However, this finding, to the best of our knowledge, is the first to indicate that emotion regulation (as indexed by RSA change) measured exclusively during nonspeech conditions (i.e., viewing of video clips) is associated with increased stuttering frequency for CWS. Contrary to predictions, CWNS also exhibited an association between emotion regulation during video viewing and frequency of stuttered disfluencies (albeit smaller in absolute terms than CWS). Thus, the speech-language planning and production of CWNS also appears vulnerable to interference associated with physiological aspects of emotion regulation.

From Porges's (2007) polyvagal perspective, decrease of RSA is associated with the recruitment of physiological resources (e.g., sympathetic arousal) to mobilize support for an individual's actively engaging with his/her environment. However, the degree or strength of physiological response that is adaptive may be task dependent and based on contextual/situational elements. For instance, when faced with extreme danger, a rapid and large autonomic response is adaptive to support the physiological resources for a fight/flight response (i.e., sympathetic preparation for action). However, during viewing of emotionally arousing video clips in a safe environment (i.e., with friendly researchers and caregivers nearby in an adjoining room), a mild-to-moderate mobilization response may be more adaptive. In the present study, it is possible that both CWS and CWNS with greater decrease of RSA during nonspeech emotionally arousing tasks are less prepared to engage in social communication. Further, when young children with greater decrease of RSA do engage socially, they may be predisposed to physiological responses during speech that do not facilitate fluent speech-language planning and production.

Hypothesis 3: For CWS, Compared to CWNS, Greater Decreases in RSA During Speaking Would Be Significantly **Associated With Increased Stuttering**

For both young CWS and their CWNS peers, there was a negative association between stuttered disfluencies and RSA change during the three speaking conditions. This finding indicates that greater decrease in RSA during speech is associated with increased stuttered disfluencies for young CWS as well as young CWNS.

As was the case with the results pertaining to the second hypothesis, these findings support our initial predictions that for young CWS, physiological aspects of emotion regulation are associated with stuttering. Again, these results are consistent with studies using behavioral measures of emotion that have reported that decreased emotion regulation during speech is associated with increased stuttering (Arnold et al., 2011; Jones, Conture, et al., 2014; Kraft et al., 2014; Walden et al., 2012). However, contrary to our initial predictions, CWNS also exhibited an association between RSA change during speaking and stuttered disfluencies (albeit smaller in absolute terms than CWS).

Both theory (Porges, 2007) and empirical evidence (e.g., Bar-Haim et al., 2004; Heilman et al., 2008; Marcovitch et al., 2010; Utendale et al., 2014) indicate that decrease in RSA is adaptive during challenging situations. However, as we previously discussed regarding the second hypothesis. the degree and directionality of change in RSA that facilitates adaptive performance may be somewhat task dependent. During social-communicative situations associated with emotional stimuli, a mild-to-moderate mobilization response may be more adaptive. Certainly, an increase in autonomic arousal from baseline (which would be associated with a greater decrease in RSA) would seem necessary to support the processes associated with speech-language planning and production in children and adults who stutter (Arnold, MacPherson, & Smith, 2014; Jones, Buhr, et al., 2014).

Comparatively less decrease, maintenance, or increase of RSA during speaking tasks may index a greater degree of emotion regulation and hence facilitate an individual's ability to engage in social communication as well as increase and/or maintain speech fluency. Children who stutter, when compared to their CWNS peers, exhibit greater emotion reactivity and lower regulation as indexed by caregiver report and behavioral (for review, see Jones, Choi, et al., 2014) and physiological measures (e.g., lower baseline RSA; Jones, Buhr, et al., 2014). Thus, it is guite possible that CWS, relative to CWNS, are at higher risk of initiating and/or maintaining a physiological response that is less supportive of fluent speech-language and more supportive of an emotionally reactive, fight/flight response.

Lack of Effect of Emotion Condition (Hypotheses 1, 2, and 3)

Contrary to our predictions for Hypotheses 1, 2, and 3. there was no effect of emotion condition on the association between stuttered disfluencies and RSA (baseline RSA and RSA change during the emotionally arousing video viewing or narrative speaking tasks). One possible explanation for the lack of effect of condition (i.e., neutral vs. negative vs. positive video clips) on young children's frequency of stuttering is that speaking represented a greater challenge than did the emotional videos, thereby minimizing the effect of the emotion conditions.

Hypothesis 4: The Stuttering of CWS With Lower Executive Functions Would Be Impacted by Physiological State (i.e., Change in RSA) More Than Those With Higher Executive Functions, and This Association Would Significantly Differ From That of CWNS

Consistent with predictions, CWS with lower executive functions exhibited a stronger association between stuttering and RSA change during speaking. However, contrary to predictions, CWNS with higher executive functions exhibited a stronger association between stuttering and RSA change during speaking. For CWS with lower executive functions, the speech-language system seems more sensitive to concurrent physiological processes associated

with emotion regulation. However, CWS with relatively well-developed executive functions appear better able to allocate cognitive resources to the initiation and/or maintenance of speech regardless of concomitant physiological activity. For these children, cognitive regulation/executive functions may serve as a "buffer," insulating or protecting their speech-language planning and production system from interference associated with concurrent physiological activity.

For CWNS, present results are somewhat more challenging to interpret. One possibility is that CWNS with higher levels of executive functions may lead to "overmonitoring" of their speech-language planning and production, which may in turn lead to increased disruptions in speech fluency when paired with greater decrease of RSA. Essentially, attempts to over-monitor, when paired with decreased emotion regulation, may lead to a dyssynchrony between top-down executive control processes and bottom-up physiological processes that does not facilitate fluent speech-language planning and/or production.

From a theoretical perspective, results for CWS support Conture and Walden's (2012) speculation that cognitive processes may serve as a link between emotions and stuttering (for further discussion of cognitive processes and stuttering, see Bosshardt, 2006; Eggers et al., 2012, 2013; Felsenfeld et al., 2010). Specifically, for CWS with higher emotion reactivity and lower regulation, cognitive resources may be diverted from concurrent speech-language planning and production processes. As Engels et al. (2007) suggested, "attention is captured by ambiguous, emotional or threatening stimuli. ... Anxiety also impairs performance on tasks that are difficult or stressful, because worrisome thoughts interfere with attention to task-relevant information. ... An implication is that, if someone is anxious, their attention is at constant risk for diversion from the task at hand, slowing or impairing performance" (p. 352). Consistent with this suggestion, Ledoux (2015) notes, "threats ... capture attention and direct it to the threat. ... Numerous laboratory studies confirm the intuition that threats or other 'emotional' stimuli can seize attention" (p. 217). Thus, emotionally arousing situations, both positive and negative in nature, may divert attention away from ongoing speechlanguage planning and production and onto real or perceived challenges, opportunities or threats, increasing the likelihood of stuttering for CWS.

As Alm (2014) suggested, it is possible that there may be a subset of CWS whose speech is related to inattention and/or attention disorders. Thus, the CWS with relatively low executive functions in the present study may represent a subgroup of CWS whose stuttering is appreciably influenced by attention processes. Of course, difficulties with cognitive processes do not rule out the possibility that physiological activity associated with emotion regulation may also be involved with instances of stuttering for these children. Thus, both cognitive and physiological processes—both being associated with temperamental reactivity and regulation—may jointly or singly contribute in complex ways to the onset, development, and maintenance of childhood stuttering.

Future empirical studies using a variety of measures of executive functions (e.g., inhibitory control, attention) should help elucidate such possibilities.

General Implications of Findings Regarding Physiological Indices of Emotion and **Childhood Stuttering**

The present findings provide evidence of an association between RSA change, executive functions, and childhood stuttering. We speculated that children's emotions may divert cognitive processes away from ongoing speechlanguage processes and thus increase the likelihood of stuttering. It is also possible that physiological activity associated with emotion regulation may directly impact speech motor control. Kleinow and Smith (2006) reported that high levels of autonomic arousal impacted speech-motor control in both adults and children. Given that some CWS exhibit less well developed speech-motor control than CWNS (e.g., MacPherson & Smith, 2012; Olander, Smith, & Zelaznik, 2010; Smith, Goffman, Sasisekaran, & Weber-Fox, 2012), it is possible that CWS's speech-motor control is more vulnerable to greater decrease in physiological emotion regulation during speech-language planning and production.

The Polyvagal Theory (Porges, 2007) provides a theoretical framework describing the potential impact of autonomic nervous system activity on vocalizations. For example, laryngeal and cardiac activity may covary, given that both the larynx and the heart receive innervation through myelinated vagal pathways originating in the nucleus ambiguus. Thus, a child with lesser cardiac regulation via the vagus, indexed by RSA change, may also display lesser laryngeal regulation via vagal pathways, with both regulatory efforts potentially contributing to difficulties initiating and/or maintaining fluent speech-language planning and production. These possibilities may be related to observations that during fluent speech, preschool-age CWS exhibit less stable (i.e., greater vocal shimmer; Hall & Yairi, 1992) as well as atypical laryngeal activity associated with consonant-vowel and vowel-consonant transitions (Conture, Rothenberg, & Molitor, 1986). However, it is speculative that RSA change may serve as an indicator of the vagal mechanisms related to laryngeal regulation in childhood stuttering, raising questions that only further empirical study can answer.

Directionality of Effect. In a recent overview, Conture et al. (2013) discussed four possible accounts of the relation between temperament, emotion and speech-language disorders. Such accounts included: (a) the (pre)dispositional model (e.g., emotions cause stuttering) (b) the disability model (e.g., stuttering causes emotions), (c) the bidirectional model (e.g., stuttering and emotion processes covary), and (d) the correlational model (e.g., stuttering and emotion are both related to a third variable such as gender). In theory, there are several ways that stuttering could impact RSA change during speaking (e.g., disability model); for example, more stuttering may lead to greater, more pervasive

disruptions in emotional regulation and hence changes in RSA from baseline. However, present findings suggest that this may not be the case for all CWS, given that CWS with relatively higher executive functions did not exhibit a significant association between RSA change and stuttering. Whether or not stuttering influences RSA change during speaking, it should be noted that the association of stuttering during narratives and RSA change during a nonspeaking task (i.e., emotionally arousing video viewing conditions) cannot be explained by emotional reactions to concurrent difficulties with speech. In this case, it is possible that decrease of RSA from baseline during emotionally arousing video viewing predisposes an individual to be at greater risk for difficulties with speech-language planning and production (e.g., predispositional model), in this case during the narrative speaking task.

A final possibility, seemingly relating to the disability model mentioned above, is that young CWS who exhibit higher, versus those with lower, stuttering frequency may have developed a general anticipation of stuttering and/or the difficulty of the upcoming speaking task. Perhaps such anticipation impacts RSA change during the preceding emotionally arousing video viewing tasks. The degree and nature of such anticipation—and whether such activity is unconscious or conscious in nature—in young CWS at or near the onset of stuttering is less well understood than in the adult population (e.g., Alm, 2004; Bowers, Saltuklaroglu, & Kalinowski, 2012; Brocklehurst, Lickley, & Corley, 2013; Sheehan, 1953). However, extant evidence suggests that CWS as young as 2 years of age have shown awareness of stuttering and/or difficulty with speech (e.g., Ambrose & Yairi, 1994; Clark, Conture, Frankel, & Walden, 2012; Ezrati-Vinacour, Platzky, & Yairi, 2001). Thus, present findings do not unequivocally support or refute any one of the aforementioned four models (e.g., bidirectional model) that attempt to account for the directionality of the association between emotion and stuttering. Instead, further empirical study (e.g., the impact of anticipation of speech and/or stuttering on RSA change in young CWS) will be necessary to establish a more comprehensive understanding of this association.

Limitations

The present study measured RSA change, a commonly used index of PNS activity, but not sympathetic nervous system activity. Beauchaine (2001) suggests that the sympathetic nervous system (SNS) should be considered simultaneously with PNS activity to establish a comprehensive account of ANS activity. For example, Jones. Buhr, et al. (2014) reported that although it is typical for SNS and PNS activity to act reciprocally (when one is high the other is low), the preschool-age CWS they studied exhibited patterns of coactivation (both high sympathetic and high parasympathetic activity) and coinhibition (low sympathetic and low parasympathetic activity) during speech. The present study did not measure whether these patterns of simultaneous ANS activity are associated with

stuttering frequency. Thus, it is suggested that future studies in this area concurrently measure SNS and PNS activity relative to frequency of (non)stuttered disfluencies (and executive functions) to more comprehensively understand the association between stuttering and PNS and SNS activity.

The present study used caregiver report to assess executive functions. Perhaps greater specificity regarding the association between RSA change and stuttering could be developed using laboratory real-time assessments of executive functions, such as inhibitory control and attention. Of course, there is still value in understanding whether and how more stable, trait-like aspects of executive functions are related to RSA change and stuttering. As suggested by Henderson and Wachs (2007), although "parent report measures do contain some subjective parental components, available evidence indicates that these measures also contain a substantial objective component that does accurately assess children's individual characteristics" (p. 402).

Conclusions

The present study provides evidence of an association between physiological aspects of young children's emotion regulation and stuttering, and how this association may be related to children's executive functions. As suggested by the Polyvagal Theory (e.g., Porges, 2007), social-communicative situations may be facilitated by parasympathetic activity supporting sufficient mobilization of resources to facilitate communication, but less marked mobilization than is associated with fight/flight responses. Further, aspects of executive functions (i.e., the composite of inhibitory control and attentional focusing used in the present study) appear to constitute an important moderator on the association between emotion regulation (as indexed by RSA change during speaking) and frequency of stuttering. Thus, executive functions may serve as one link between physiological associates of emotion regulation and the difficulties these children have establishing normally fluent speech.

Certainly, one cannot categorically ignore the possibility that experience with, as well as learned reactions to stuttering, impacts the behaviors, characteristics, and performance of individuals who stutter, even young children. In this case, the behaviors, characteristics, and so on, are associated with emotional processes. Of course, much the same could be said about empirical studies of cognitive, learned, linguistic, and motoric contributions to childhood stuttering. However, given that present participants were at or near the onset of stuttering, these participants had fewer opportunities to experience/learn reactions to stuttering than older children, teens, and adults who stutter. Thus, it is difficult to readily or completely attribute this study's cognitive, physiological, and other findings to these young children's experience with/learned reactions to stuttering. Instead, cognitive, emotional, and related processes appear to play a meaningful role in the onset and/or developmental trajectory of childhood stuttering,

a possibility that warrants consideration as part of any comprehensive account of childhood stuttering.

Acknowledgments

This study would not have been possible without financial support from the following: National Institutes of Health (NIH) grants from the National Institute on Deafness and Other Communication Disorders (NIDCD) to Vanderbilt University (5R01DC000523-19, 2R56DC000523-20A1), the National Center for Research Resources, and a CTSA grant (1 UL1 RR024975-01) to Vanderbilt University that is now at the National Center for Advancing Translational Sciences (UL1 TR000445-06). The research reported herein does not reflect the views of the NIH, NIDCD, or Vanderbilt University. The authors thank Kristopher J. Preacher for his advice and comments on data analysis and visual display of statistical analyses. The authors would like to extend their sincere appreciation for reviews of early versions of this article by Dr. Julie Liss, Editor, Dr. Hans-Georg Bosshardt, Associate Editor, and two anonymous reviewers. The authors also extend sincere appreciation to the young children and their caregivers who participated in this study, individuals without whose cooperation this project would not have been possible to conduct.

References

- Aiken, L. S., Mistler, S. A., Coxe, S., & West, S. G. (2015). Analyzing count variables in individuals and groups: Single level and multilevel models. Group Processes & Intergroup Relations, 1368430214556702. https://doi.org/10.1177/1368430214556702
- Aiken, L. S., & West, S. G. (1991). Multiple regression: Testing and interpreting interactions. Thousand Oaks, CA: Sage.
- Alm, P. A. (2004). Stuttering, emotions, and heart rate during anticipatory anxiety: A critical review. Journal of Fluency Disorders, 29, 123-133. https://doi.org/10.1016/j.jfludis.2004.02.001
- Alm, P. A. (2014). Stuttering in relation to anxiety, temperament, and personality: Review and analysis with focus on causality. Journal of Fluency Disorders, 40, 5-21. https://doi.org/10.1016/ j.jfludis.2014.01.004
- Ambrose, N. G., & Yairi, E. (1994). The development of awareness of stuttering in preschool-children. Journal of Fluency Disorders, 19, 229-245.
- Anderson, J. D., Pellowski, M. W., Conture, E. G., & Kelly, E. M. (2003). Temperamental characteristics of young children who stutter. Journal of Speech, Language, and Hearing Research, 46, 1221–1233. https://doi.org/10.1044/1092-4388(2003/095)
- Arnold, H. S., Conture, E. G., Key, A. P. F., & Walden, T. (2011). Emotional reactivity, regulation and childhood stuttering: A behavioral and electrophysiological study. Journal of Communication Disorders, 44(3), 276-293. https://doi.org/10.1016/ j.jcomdis.2010.12.003
- Arnold, H. S., MacPherson, M. K., & Smith, A. (2014). Autonomic correlates of speech versus nonspeech tasks in children and adults. Journal of Speech, Language, and Hearing Research, 57(4), 1296-1307. https://doi.org/10.1044/2014_JSLHR-S-12-0265
- Atkins, D. C., Baldwin, S. A., Zheng, C., Gallop, R. J., & Neighbors, C. (2013). A tutorial on count regression and zero-altered count models for longitudinal substance use data. Psychology of Addictive Behaviors: Journal of the Society of Psychologists in Addictive Behaviors, 27(1), 166-177. https://doi.org/10.1037/ a0029508
- Bar-Haim, Y., Fox, N. A., VanMeenen, K. M., & Marshall, P. J. (2004). Children's narratives and patterns of cardiac reactivity.

- Developmental Psychobiology, 44, 238-249. https://doi.org/ 10.1002/dev.20006
- Beauchaine, T. (2001). Vagal tone, development, and Gray's motivational theory: Toward an integrated model of autonomic nervous system functioning in psychopathology. Development and Psychopathology, 13(2), 183-214.
- Beauchaine, T., Gatzke-Kopp, L., & Mead, H. K. (2007). Polyvagal theory and developmental psychopathology: Emotion dysregulation and conduct problems from preschool to adolescence. Biological Psychology, 74, 174-184. https://doi.org/ 10.1016/j.biopsycho.2005.08.008
- Becker, D. R., Carrère, S., Siler, C., Jones, S., Bowie, B., & Cooke, C. (2012). Autonomic regulation on the stroop predicts reading achievement in school age children. Mind, Brain, and Education, 6(1), 10–18. https://doi.org/10.1111/j.1751-228X. 2011.01130.x
- Bosshardt, H. G. (2006). Cognitive processing load as a determinant of stuttering: Summary of a research programme. Clinical Linguistics & Phonetics, 20, 371-385. https://doi.org/10.1080/ 02699200500074321
- Bowers, A., Saltuklaroglu, T., & Kalinowski, J. (2012). Autonomic arousal in adults who stutter prior to various reading tasks intended to elicit changes in stuttering frequency. International Journal of Psychophysiology, 83(1), 45-55. https://doi.org/ 10.1016/j.ijpsycho.2011.09.021
- Brocklehurst, P. H., Lickley, R. J., & Corley, M. (2013). Revisiting Bloodstein's anticipatory struggle hypothesis from a psycholinguistic perspective: A variable release threshold hypothesis of stuttering. Journal of Communication Disorders, 46(3), 217-237. https://doi.org/10.1016/j.jcomdis. 2013.04.002
- Cabanac, M. (2002). What is emotion? Behavioural Processes, 60(2), 69-83. https://doi.org/10.1016/S0376-6357(02)00078-5
- Calkins, S. D. (1997). Cardiac vagal tone indices of temperamental reactivity and behavioral regulation in young children. Developmental Psychobiology, 31, 125-135.
- CardioBatch. (2007). Brain-body center. Chicago, IL: University of Illinois at Chicago.
- CardioEdit. (2007). Brain-body center. Chicago, IL: University of Illinois at Chicago.
- Carlson, S. M., & Wang, T. S. (2007). Inhibitory control and emotion regulation in preschool children. Cognitive Development, 22(4), 489-510. https://doi.org/10.1016/j.cogdev.2007. 08.002
- Chapman, H. A., Woltering, S., Lamm, C., & Lewis, M. D. (2010). Hearts and minds: Coordination of neurocognitive and cardiovascular regulation in children and adolescents. Biological Psychology, 84(2), 296-303. https://doi.org/10.1016/ j.biopsycho.2010.03.001
- Choi, D., Conture, E. G., Walden, T. A., Jones, R. M., & Kim, H. (2016). Emotional diathesis, emotional stress, and childhood stuttering. Journal of Speech, Language, and Hearing Research, 59(4), 616-630. https://doi.org/10.1044/2015_JSLHR-S-14-0357
- Choi, D., Conture, E. G., Walden, T. A., Lambert, W. E., & Tumanova, V. (2013). Behavioral inhibition and childhood stuttering. Journal of Fluency Disorders, 38(2), 171-183. https://doi.org/10.1016/j.jfludis.2013.03.001
- Clark, C. E., Conture, E. G., Frankel, C. B., & Walden, T. A. (2012). Communicative and psychological dimensions of the KiddyCAT. Journal of Communication Disorders, 45(3), 223–234. https://doi.org/10.1016/j.jcomdis.2012.01.002
- Clark, C. E., Conture, E. G., Walden, T. A., & Lambert, W. E. (2015). Speech-language dissociations, distractibility, and childhood stuttering. American Journal of Speech-Language

- Pathology, 24(3), 480–503. https://doi.org/10.1044/2015_ AJSLP-14-0198
- Cohen, J. (1988). Statistical power analysis for the behavioral sciences (2nd ed.). Hillsdale, NJ: Routledge.
- Conture, E. G. (2001). Stuttering: Its nature, diagnosis, and treatment. Boston, MA: Allyn and Bacon.
- Conture, E. G., Kelly, E. M., & Walden, T. A. (2013). Temperament, speech and language: An overview. Journal of Communication Disorders, 46(2), 125–142. https://doi.org/10.1016/j.jcomdis. 2012.11.002
- Conture, E. G., Rothenberg, M., & Molitor, R. D. (1986). Electroglottographic observations of young stutterers' fluency. Journal of Speech and Hearing Research, 29, 384-393.
- Conture, E. G., & Walden, T. A. (2012). Dual diathesis-stressor model of stuttering. In L. Beliakova & Y. Filatova (Eds.), Theoretical issues of fluency disorders (pp. 94-127). Moscow, Russia: Vlados.
- Denver, J. W., Reed, S. F., & Porges, S. W. (2007). Methodological issues in the quantification of respiratory sinus arrhythmia. Biological Psychology, 74, 286-294. https://doi.org/10.1016/ j.biopsycho.2005.09.005
- Derryberry, D., & Reed, M. A. (2002). Anxiety-related attentional biases and their regulation by attentional control. Journal of Abnormal Psychology, 111(2), 225-236. https://doi.org/ 10.1037//0021-843X.111.2.225
- Dunn, L., & Dunn, L. (2007). Peabody Picture Vocabulary Test-Fourth Edition (PPVT-4). Minneapolis, MN: AGS.
- Eggers, K., De Nil, L. F., & Van den Bergh, B. R. (2010). Temperament dimensions in stuttering and typically developing children. Journal of Fluency Disorders, 35, 355-372. https://doi. org/10.1016/j.jfludis.2010.10.004
- Eggers, K., De Nil, L. F., & Van den Bergh, B. R. H. (2012). The efficiency of attentional networks in children who stutter. Journal of Speech, Language, and Hearing Research, 55(3), 946-959. https://doi.org/10.1044/1092-4388(2011/10-0208)
- Eggers, K., De Nil, L. F., & Van den Bergh, B. R. H. (2013). Inhibitory control in childhood stuttering. Journal of Fluency Disorders, 38(1), 1-13. https://doi.org/10.1016/j.jfludis.2012.10.001
- El-Sheikh, M. (2005). Stability of respiratory sinus arrhythmia in children and young adolescents: A longitudinal examination. Developmental Psychobiology, 46(1), 66-74. https://doi.org/ 10.1002/dev.20036
- Engels, A. S., Heller, W., Mohanty, A., Herrington, J. D., Banich, M. T., Webb, A. G., & Miller, G. A. (2007). Specificity of regional brain activity in anxiety types during emotion processing. Psychophysiology, 44(3), 352-363. https://doi.org/10.1111/ j.1469-8986.2007.00518.x
- Ezrati-Vinacour, R., Platzky, R., & Yairi, E. (2001). The young child's awareness of stuttering-like disfluency. Journal of Speech, Language, and Hearing Research, 44, 368-380.
- Felsenfeld, S., van Beijsterveldt, C. E. M., & Boomsma, D. I. (2010). Attentional regulation in young twins with probable stuttering, high nonfluency, and typical fluency. Journal of Speech, Language, and Hearing Research, 53, 1147-1166. https://doi.org/10.1044/1092-4388(2010/09-0164)
- Gardner, W., Mulvey, E. P., & Shaw, E. C. (1995). Regression analyses of counts and rates: Poisson, overdispersed Poisson, and negative binomial models. Psychological Bulletin, 118(3), 392-404. https://doi.org/10.1037/0033-2909.118.3.392
- Gentzler, A. L., Santucci, A. K., Kovacs, M., & Fox, N. A. (2009). Respiratory sinus arrhythmia reactivity predicts emotion regulation and depressive symptoms in at-risk and control children. Biological Psychology, 82(2), 156-163. https://doi.org/10.1016/ j.biopsycho.2009.07.002

- Gerardi-Caulton, G. (2000). Sensitivity to spatial conflict and the development of self-regulation in children 24-36 months of age. Developmental Science, 3(4), 397-404. https://doi.org/ 10.1111/1467-7687.00134
- Gilissen, R., Koolstra, C. M., van Ijzendoorn, M. H., Bakermans-Kranenburg, M. J., & van der Veer, R. (2007). Physiological reactions of preschoolers to fear-inducing film clips: Effects of temperamental fearfulness and quality of the parent-child relationship. Developmental Psychobiology, 49, 187–195. https:// doi.org/10.1002/dev.20188
- Goldman, R., & Fristoe, M. (2000). Goldman Fristoe Test of Articulation-Second Edition. Circle Pines, MN: American Guidance Service.
- Gross, J. J. (2002). Emotion regulation: Affective, cognitive, and social consequences. Psychophysiology, 39, 281-291. https:// doi.org/10.1017/S0048577201393198
- Gross, J. J., & Thompson, R. A. (2007). Emotion regulation: Conceptual foundations. In J. Gross (Ed.), Handbook of emotion regulation (pp. 3-24). New York, NY: Guilford Press.
- Hall, K. D., & Yairi, E. (1992). Fundamental-frequency, jitter, and shimmer in preschoolers who stutter. Journal of Speech and Hearing Research, 35, 1002-1008.
- Hastings, P. D., Kahle, S., & Nuselovici, J. M. (2014). How well socially wary preschoolers fare over time depends on their parasympathetic regulation and socialization. Child Development, 85(4), 1586-1600. https://doi.org/10.1111/cdev. 12228
- Heilman, K. J., Bal, E., Bazhenova, O. V., Sorokin, Y., Perlman, S. B., Hanley, M. C., & Porges, S. W. (2008). Physiological responses to social and physical challenges in children: Quantifying mechanisms supporting social engagement and mobilization behaviors. Developmental Psychobiology, 50(2), 171-182. https://doi.org/10.1002/dev.20257
- Henderson, H. A., & Wachs, T. D. (2007). Temperament theory and the study of cognition-emotion interactions across development. Developmental Review, 27(3), 396-427. https://doi.org/ 10.1016/j.dr.2007.06.004
- Hollingshead, A. (1975). Four Factor Index of Social Status. New Haven, CT: Department of Sociology, Yale University.
- Holmbeck, G. N. (2002). Post-hoc probing of significant moderational and mediational effects in studies of pediatric populations. Journal of Pediatric Psychology, 27(1), 87-96. https:// doi.org/10.1093/jpepsy/27.1.87
- Hresko, W. P., Reid, D. K., & Hammill, D. D. (1999). Test of Early Language Development-Third Edition (TELD-3). Austin, TX: Pro-Ed.
- Izard, C. E. (2010). The many meanings/aspects of emotion: Definitions, functions, activation, and regulation. Emotion Review, 2(4), 363-370. https://doi.org/10.1177/1754073910374661
- Johnson, K. N., Conture, E. G., & Walden, T. A. (2012). Efficacy of attention regulation in preschool-age children who stutter: A preliminary investigation. Journal of Communication Disorders, 45(4), 263-278. https://doi.org/10.1016/j.jcomdis.2012. 04.001
- Johnson, K. N., Karrass, J., Conture, E. G., & Walden, T. A. (2009). Influence of stuttering variation on talker group classification in preschool children: Preliminary findings. Journal of Communication Disorders, 42(3), 195-210. https://doi.org/ 10.1016/j.jcomdis.2008.12.001
- Johnson, K. N., Walden, T. A., Conture, E. G., & Karrass, J. (2010). Spontaneous regulation of emotions in preschool children who stutter: Preliminary findings. Journal of Speech, Language, and Hearing Research, 53, 1478-1495. https://doi.org/ 10.1044/1092-4388(2010/08-0150)

- Jones, R. M., Buhr, A. P., Conture, E. G., Tumanova, V., Walden, T. A., & Porges, S. W. (2014). Autonomic nervous system activity of preschool-age children who stutter. *Journal of Fluency Disorders*, 41, 12–31. https://doi.org/10.1016/j.jfludis.2014. 06.002
- Jones, R. M., Choi, D., Conture, E., & Walden, T. (2014). Temperament, emotion, and childhood stuttering. *Seminars in Speech and Language*, 35(2), 114–131. https://doi.org/10.1055/s-0034-1371755
- Jones, R. M., Conture, E. G., & Walden, T. A. (2014). Emotional reactivity and regulation associated with fluent and stuttered utterances of preschool-age children who stutter. *Journal of Communication Disorders*, 48, 38–51. https://doi.org/10.1016/ j.jcomdis.2014.02.001
- Karrass, J., Walden, T. A., Conture, E. G., Graham, C. G., Arnold, H. S., Hartfield, K. N., & Schwenk, K. A. (2006). Relation of emotional reactivity and regulation to childhood stuttering. *Journal of Communication Disorders*, 39, 402–423. https://doi.org/10.1016/J.Jcomdis.2005.12.004
- Kefalianos, E., Onslow, M., Block, S., Menzies, R., & Reilly, S. (2012). Early stuttering, temperament and anxiety: Two hypotheses. *Journal of Fluency Disorders*, 37(3), 151–163. https://doi.org/10.1016/j.jfludis.2012.03.002
- Kleinow, J., & Smith, A. (2006). Potential interactions among linguistic, autonomic, and motor factors in speech. *Developmental Psychobiology*, 48(4), 275–287. https://doi.org/10.1002/dev.20141
- Kraemer, H., Wilson, G., Fairburn, C. G., & Agras, W. (2002). Mediators and moderators of treatment effects in randomized clinical trials. *Archives of General Psychiatry*, 59(10), 877–883. https://doi.org/10.1001/archpsyc.59.10.877
- Kraft, S. J., Ambrose, N., & Chon, H. (2014). Temperament and environmental contributions to stuttering severity in children: The role of effortful control. *Seminars in Speech and Language*, 35(02), 080–094. https://doi.org/10.1055/s-0034-1371753
- Kreibig, S. D. (2010). Autonomic nervous system activity in emotion: A review. *Biological Psychology*, 84(3), 394–421. https://doi.org/10.1016/j.biopsycho.2010.03.010
- LeDoux, J. (2015). Anxious: Using the brain to understand and treat fear and anxiety (1st ed.). New York, NY: Penguin Books.
- Levelt, W. J., Roelofs, A., & Meyer, A. S. (1999). A theory of lexical access in speech production. *The Behavioral and Brain Sciences*, 22, 1–38, 38–75.
- Lewis, G. F. (2010). *RespFreqFromRSA*. Chicago, IL: Brain-Body Center, University of Illinois at Chicago.
- Lewis, G. F., Furman, S. A., McCool, M. F., & Porges, S. W. (2012). Statistical strategies to quantify respiratory sinus arrhythmia: Are commonly used metrics equivalent?. *Biological Psychology*, 89(2), 349–364. https://doi.org/10.1016/j.biopsycho. 2011.11.009
- Lonigan, C. J., & Vasey, M. W. (2008). Negative affectivity, effortful control, and attention to threat-relevant stimuli. *Journal of Abnormal Child Psychology*, 37(3), 387–399. https://doi.org/10.1007/s10802-008-9284-y
- MacPherson, M. K., & Smith, A. (2012). Influences of sentence length and syntactic complexity on the speech motor control of children who stutter. *Journal of Speech, Language, and Hearing Research*, 56(1), 89–102. https://doi.org/10.1044/1092-4388(2012/11-0184)
- Marcovitch, S., Leigh, J., Calkins, S. D., Leerks, E. M., O'Brien, M., & Blankson, A. N. (2010). Moderate vagal withdrawal in 3.5-year-old children is associated with optimal performance on executive function tasks. *Developmental Psychobiology*, 52(6), 603–608. https://doi.org/10.1002/dev.20462

- Mayer, M. (1969). Frog, where are you? New York, NY: Dial Books for Young Readers.
- McGraw, K. O., & Wong, S. P. (1996). Forming inferences about some intraclass correlation coefficients. *Psychological Methods*, 1(1), 30–46. https://doi.org/10.1037/1082-989X.1.1.30
- Nich, C., & Carroll, K. (1997). Now you see it, now you don't: A comparison of traditional versus random-effects regression models in the analysis of longitudinal follow-up data from a clinical trial. *Journal of Consulting and Clinical Psychology*, 65(2), 252–261. https://doi.org/http://dx.doi.org/10.1037/0022-006X.65.2.252
- Ntourou, K., Conture, E. G., & Walden, T. A. (2013). Emotional reactivity and regulation in preschool-age children who stutter. *Journal of Fluency Disorders*, *38*(3), 260–274. https://doi.org/10.1016/j.jfludis.2013.06.002
- Olander, L., Smith, A., & Zelaznik, H. N. (2010). Evidence that a motor timing deficit is a factor in the development of stuttering. *Journal of Speech, Language, and Hearing Research*, 53, 876–886. https://doi.org/10.1044/1092-4388(2009/09-0007)
- Osborne, J. W., & Overbay, A. (2004). The power of outliers (and why researchers should always check for them). *Practical Assessment, Research & Evaluation*, 9(6). Retrieved from http://PAREonline.net/getvn.asp?v=9&n=6
- Peers, P. V., Simons, J. S., & Lawrence, A. D. (2013). Prefrontal control of attention to threat. Frontiers in Human Neuroscience, 7. https://doi.org/10.3389/fnhum.2013.00024
- Pinheiro, J. C., & Bates, D. M. (2013). Mixed-effects models in S and S-PLUS. New York, NY: Springer.
- Porges, S. W. (1985). U.S. Patent No. 4510944A. Washington, DC: U.S. Patent and Trademark Office.
- **Porges, S. W.** (1992). Vagal tone: A physiologic marker of stress vulnerability. *Pediatrics*, *90*(3), 498–504.
- **Porges, S. W.** (2001). The polyvagal theory: Phylogenetic substrates of a social nervous system. *International Journal of Psychophysiology: Official Journal of the International Organization of Psychophysiology*, 42, 123–146.
- Porges, S. W. (2007). The polyvagal perspective. Biological Psychology, 74, 116–143. https://doi.org/10.1016/j.biopsycho.2006. 06.009
- Porges, S. W., & Bohrer, R. E. (1990). Analyses of periodic processes in psychophysiological research. In J. T. Cacioppo & L. G. Tassinary (Eds.), *Principles of psychophysiology: Physical, social, and inferential elements* (pp. 708–753). New York, NY: Cambridge University Press.
- Porges, S. W., Doussard-Roosevelt, J. A., Portales, A. L., & Greenspan, S. I. (1996). Infant regulation of the vagal "brake" predicts child behavior problems: A psychobiological model of social behavior. *Developmental Psychobiology*, 29, 697–712. https://doi.org/10.1002/(SICI)1098-2302(199612)29:8<697:: AID-DEV5>3.0.CO;2-O
- Razali, N. M., & Wah, Y. B. (2011). Power comparisons of Shapiro-Wilk, Kolmogorov-Smirnov, Lilliefors and Anderson-Darling tests. *Journal of Statistical Modeling and Analytics*, 2(1), 21–33.
- Riley, G. D. (1994). Stuttering severity instrument for children and adults (3rd ed.). Austin, TX: Pro-Ed.
- Rothbart, M. K., Ahadi, S. A., & Evans, D. E. (2000). Temperament and personality: Origins and outcomes. *Journal of Personality and Social Psychology*, 78, 122–135.
- Rothbart, M. K., Ahadi, S. A., Hershey, K. L., & Fisher, P. (2001). Investigations of temperament at three to seven years: The Children's Behavior Questionnaire. *Child Development*, 72(5), 1394–1408. https://doi.org/10.1111/1467-8624.00355
- Schwenk, K. A., Conture, E. G., & Walden, T. A. (2007). Reaction to background stimulation of preschool children who do and do

- not stutter. Journal of Communication Disorders, 40, 129-141. https://doi.org/10.1016/j.jcomdis.2006.06.003
- Sheehan, J. G. (1953). Theory and treatment of stuttering as an approach-avoidance conflict. The Journal of Psychology, 36(1), 27-49. https://doi.org/10.1080/00223980.1953.9712875
- Shrout, P. E., & Fleiss, J. L. (1979). Intraclass correlations: Uses in assessing rater reliability. Psychological Bulletin, 86(2),
- Singer, J. D., & Willett, J. B. (2003). Applied longitudinal data analysis: Modeling change and event occurrence (1st ed.). Oxford, England; New York, NY: Oxford University Press.
- Smith, A., Goffman, L., Sasisekaran, J., & Weber-Fox, C. (2012). Language and motor abilities of preschool children who stutter: Evidence from behavioral and kinematic indices of nonword repetition performance. Journal of Fluency Disorders, 37(4), 344–358. https://doi.org/10.1016/j.jfludis.2012.06.001
- Sonnemans, J., & Frijda, N. H. (1994). The structure of subjective emotional intensity. Cognition & Emotion, 8(4), 329-350. https://doi.org/10.1080/02699939408408945
- Staton, L., El-Sheikh, M., & Buckhalt, J. A. (2009). Respiratory sinus arrhythmia and cognitive functioning in children. Developmental Psychobiology, 51(3), 249-258. https://doi.org/10.1002/dev.20361
- Tumanova, V., Conture, E. G., Lambert, E. W., & Walden, T. A. (2014). Speech disfluencies of preschool-age children who do and do not stutter. Journal of Communication Disorders, 49, 25-41. https://doi.org/10.1016/j.jcomdis.2014.01.003
- Utendale, W. T., Nuselovici, J., Saint-Pierre, A. B., Hubert, M., Chochol, C., & Hastings, P. D. (2014). Associations between inhibitory control, respiratory sinus arrhythmia, and externalizing problems in early childhood. Developmental Psychobiology, 56(4), 686-699. https://doi.org/10.1002/dev.21136
- Walden, T. A., Frankel, C. B., Buhr, A. P., Johnson, K. N., Conture, E. G., & Karrass, J. M. (2012). Dual diathesis-stressor model of emotional and linguistic contributions to developmental stuttering. Journal of Abnormal Child Psychology, 40(4), 633-644. https://doi.org/10.1007/s10802-011-9581-8

- Weber, E. J. M., Van Der Molen, M. W., & Molenaar, P. C. M. (1994). Heart rate and sustained attention during childhood: Age changes in anticipatory heart rate, primary bradycardia, and respiratory sinus arrhythmia. Psychophysiology, 31(2), 164-174. https://doi.org/10.1111/j.1469-8986.1994.tb01036.x
- White, L. K., Helfinstein, S. M., Reeb-Sutherland, B. C., Degnan, K. A., & Fox, N. A. (2009). Role of attention in the regulation of fear and anxiety. Developmental Neuroscience, 31(4), 309-317. https://doi.org/10.1159/000216542
- White, L. K., McDermott, J. M., Degnan, K. A., Henderson, H. A., & Fox, N. A. (2011). Behavioral inhibition and anxiety: The moderating roles of inhibitory control and attention shifting. Journal of Abnormal Child Psychology, 39(5), 735-747. https:// doi.org/10.1007/s10802-011-9490-x
- Wilder, J. (1962). Basimetric approach (law of initial value) to biological rhythms. Annals of the New York Academy of Sciences, 98(4), 1211–1220. https://doi.org/10.1111/j.1749-6632.1962.tb30629.x
- Williams, K. (2007). Expressive Vocabulary Test-Second Edition (EVT-2). Circle Pines, MN: AGS.
- Yaruss, J. S. (1997). Clinical implications of situational variability in preschool children who stutter. Journal of Fluency Disorders, 22(3), 187-203. https://doi.org/10.1016/S0094-730X(97)00009-0
- Yaruss, J. S. (1998). Real-time analysis of speech fluency: Procedures and reliability training. American Journal of Speech-Language Pathology, 7(2), 25-37.
- Zeger, S. L., & Liang, K.-Y. (1986). Longitudinal data analysis for discrete and continuous outcomes. Biometrics, 42(1), 121-130. https://doi.org/10.2307/2531248
- Zeger, S. L., Liang, K.-Y., & Albert, P. S. (1988). Models for longitudinal data: A generalized estimating equation approach. Biometrics, 44(4), 1049-1060. https://doi.org/10.2307/2531734
- Zengin-Bolatkale, H., Conture, E. G., & Walden, T. A. (2015). Sympathetic arousal of young children who stutter during a stressful picture naming task. Journal of Fluency Disorders, 46, 24-40. https://doi.org/10.1016/j.jfludis.2015.07.005